Title:
A rare case of renal oncocytoma associated with erythrocytosis: Case report

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Background

Erythrocytosis occurs as a paraneoplastic syndrome in 3.5% of patients with renal cell carcinoma. Polycythemia is usually attributed to erythropoietin production by the tumor or by the adjacent normal parenchyma in response to hypoxia induced by tumor growth.\(^1\) Although cases of concomitant polycythemia have been reported for a variety of urologic tumors, there are no reports about a possible association with renal oncocytomas. In this case report we present a unique case of renal oncocytoma associated with polycythemia.

Case Report

A 41-year-old man presented in March 2005 to the internist due to frequent headaches and plethoric face. The CBC results showed a hematocrit of 65.1%. Hemoglobin was 21.7 g/dl, with all other hematological, biochemical and urine examinations being normal, apart from a small cholesterol (232mg/dl) and GPT (72U/L) level elevation. The patient was taking no medications and his medical and surgical history was clear.

Findings of the abdominal ultrasound examination were normal, apart from a mass on the lower pole of the right kidney, measuring 8.1 x 6.1cm, with calcifications. On subsequent CT and MRI examination the lower pole mass measured 6 x 6 x 7cm and the differential diagnosis included oncocytoma and renal cell carcinoma (Figure 1). There was evidence of a tumor pseudocapsule with similar uptake to that of the normal renal parenchyma and a central part showing homogeneously diminished contrast uptake. Furthermore, the mass appeared to distort the lower pole calyces of the right kidney. No
renal vein invasion or lymph node enlargement was noted. Erythropoietin level was 17.4 mIU/ml (normal range: 9-26 mIU/ml).

Subsequently, after phlebotomy (twice), the patient was subjected to open partial nephrectomy via a flank approach. Macroscopically, the tumor demonstrated a brownish color. It was solid, well circumscribed and showed no areas of necrosis. A central scar was apparent on macroscopic examination. Microscopically, the tumor showed the morphological and immunohistochemical characters of oncocytoma (vimentin (-), CK7 (-), CD (+)).

The hematocrit levels dropped to normal levels in the perioperative period (probably due to phlebotomy and blood loss during the operation). The patient was discharged from the hospital after an uneventful recovery period. On follow-up examination 1, 3 and 6 months later the hematocrit remained normal (42 to 43%, hemoglobin 14-14.5g/dl) and after 9 months the hematocrit was 42.5%. Follow-up MRI at 9 months (Figure 2) was normal and erythropoietin levels dropped to 14.5 mIU/ml one year after the operation.

Discussion

About 7% of surgically excised renal neoplasms are oncocytomas.\(^2\) Most are asymptomatic at presentation and are discovered incidentally during evaluation for nonurological problems, whereas hematuria and pain occur in a minority of patients. Radiologically, a central scar is often found on ultrasound and CT, however, this is considered nonspecific and occurs in only 33% of oncocytomas.\(^2\) Because of the lack of
pathognomonic radiographic signs, the diagnosis of oncocytoma is rarely made without operative exploration.

Paraneoplastic phenomena have not been reported for renal oncocytomas. However, paraneoplastic manifestations are present in up to 20% of renal cell carcinomas, sometimes being the first clinical presentation, and most of them subside after surgical treatment of the tumor.\(^3\) Erythropoietin has been immunolocalized to the cytoplasm of renal cell carcinoma cells in the majority of cases with clinical erythrocytosis.\(^4\) In a study of 165 renal cell carcinomas by Ljungberg \textit{et al}, 33% of renal cell carcinomas had elevated serum erythropoietin, however, no correlation between erythrocytosis and elevated serum erythropoietin was found.\(^5\)

This is the first report of an oncocytoma with associated erythrocytosis. The serum erythropoietin levels were within the normal range in this case and dropped by 17% one year after the operation, in parallel with stabilisation of the hematocrit to normal levels. The possibility that erythrocytosis was a result of the response to tumor-induced hypoxia can not be excluded. The contribution of erythropoietin to erythrocytosis in this particular case is unclear, however, the fact that the erythrocytosis was corrected after tumor resection strongly suggests that this was a tumor-related occurrence.
References


Legends

Figure 1: MRI showing a solid right lower pole mass, well circumscribed with inhomogeneous internal structure.

Figure 2: Follow-up MRI at 9 months postoperatively showing normal remaining kidney with scar formation.